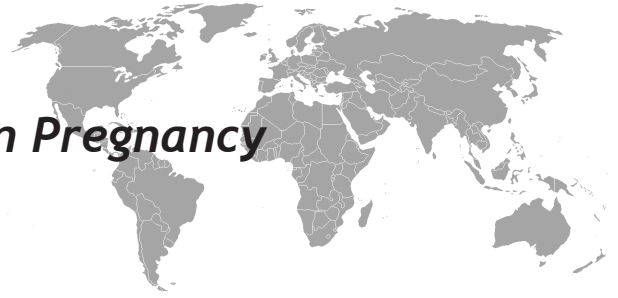


Nasal Septal Angiofibroma in Pregnancy



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ABSTRACT

Extra nasopharyngeal angiofibroma (ENA) is a term used for fibrous nodules that are located outside the nasopharynx. Location of angiofibromas outside the nasopharynx is rare. In addition, septum is an extremely rare area for involvement. While 11 nasal septum-originated cases have been reported until today in the literature, no septal angiofibromas during pregnancy have been reported yet. We are presenting a nasal septum-originated angiofibroma case in a 26-year old pregnant woman, together with literature data.

Key words: Pregnancy, extranasopharyngeal angiofibrom, benign tumours

Gebelikte Nazal Septal Anjiofibroma

ÖZET

Ekstranasofarengel anjiofibroma (ENA) nazofarenks dışında yerleşmiş fibröz nodüller için kullanılan bir terimdir. Anjiofibromlar nazofarenks dışında nadir görülürler. Nazal septum ise oldukça nadir yerleşim yerlerinden biridir. Literatürde günümüze kadar 12 nazal septum kaynaklı anjiofibroma olgusu bildirilmiştir. Fakat literatürde gebelik esnasında görülen nazal septum anjiofibroması henüz mevcut değildir. Çalışmamızda nazal septum kaynaklı 26 yaşında gebe anjiofibroma olgusu literatür verileri eşliğinde sunuldu.

Anahtar kelimeler: Gebelik, ekstranasofaringeal anjiofibrom, benign tümör

INTRODUCTION

Juvenile nasopharyngeal angiofibroma (JNA) is a neoplasia that is histologically benign, locally aggressive, non-encapsulated, extremely vascularized and commonly located in the nasopharynx (1). Although it is the most common benign tumor observed in the nasopharynx, its incidence is 0.5% among all head-neck tumors. It usually originates from the trifurcation of the palatine bone, superior margin of sphenopalatine foramen which is formed by horizontal ala of vomerine and sphenoid pterygoid process root (2). JNA may cause fatal complications, such as intracranial invasion and bleeding. Extranasopharyngeal angiofibroma (ENA) is a term used for fibrous nodules that are located outside the nasopharynx.

Clinic manifestations of extranasopharyngeal angiofibroma are very different than manifestations of nasopharyngeal angiofibroma. ENA is a very rare case and septum involvement is also extremely rare (1). We are presenting a case of 26-year old pregnant woman, who was admitted to our outpatient hospital with nasal septum originated angiofibroma, together with the literature data.

CASE

The 26-year-old female patient was admitted to our outpatient hospital in the fifth month of her pregnancy, with the complaint of epistaxis recurrences for 1 month. According to the anterior rhinoscopy, a small bleeding ul-

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cerated lesion, originated from right nasal septum, was detected. Results from punch biopsy of the mass were reported as “capillary hemangioma”. No major bleeding was observed after the biopsy. Treatment was planned after the delivery but the patient was admitted to our clinic three months later, with recurrent epistaxis and severe nasal congestion of the right side. According to the anterior rhinoscopy and endoscopy performed, a hemorrhagic mass with 2-2.5 cm in diameter, which filled almost all of right nasal passage and protruded out of the nostril, was detected. Bleeding was controlled with an anterior compress. As delivery would occur in two weeks, patient was taken to follow-up. Approximately one month after the birth, the examination of the patient revealed a mass in the right nasal cavity, which originated from frontal nasal septum and was diminished to 0.8-1 cm. No protrusion of mass into nasopharynx or paranasal sinuses was present. Routine laboratory assessments were unremarkable except anemia. The mass was removed enblock, with mucosa and periost, under local anesthesia. Intra-operative bleeding was minimal. Reported histopathology result stated angiofibroma (Figure 1). No residual or recurrent sign was detected in post-operative examinations.

DISCUSSION

JNA is a rare benign tumor, which usually exists in men. Angiofibromas are rarely seen in regions other than the nasopharynx. ENA of head and neck is much rarer than JNA (1,3,4). Windfuhr and Remmert have reported 65 ENA cases in their review study (3). Most often, they originate from the maxillary sinus (24.6%), but there are cases reported to originate from regions other than the maxillary sinus, such as the ethmoid sinus, nasal cavity, septum, larynx, cheek, conjunctiva, oropharynx, tonsil, retro molar area, middle and inferior turbinate (1-14). Septum originated angiofibroma is extremely rare and 12 septum-originated cases have been reported in the literature, until today (Table 1) (1,4,6-14). This case is also the first case of septum-originated angiofibroma during pregnancy.

Numerous theories on the origins and development of angiofibromas were reported (developmental, hormonal and genetic disorders) (1,6). Hiraide and Matsubara suggested that these tumors originated from the periosteum of the perpendicular plate, which is located in the ethmoid bone where the fascia basalis is located (6). Akbas et al. suggested that these tumors originated from ecto-

pic tissue, as anterior nasal septum did not contain fascia basalis (1). We suggest that hormonal factors may have a key role in our case, the reason being that the tumor developed during pregnancy and it regressed after birth.

ENA is clinically distinctive from JNAs. Reasons for admission to hospital are generally epistaxis and slow developing nasal congestion on one side (1,3,4). In contrast to JNA, ENA is seen in elderly women, its symptoms are fast progressive, and it has less hypervascularity (3,4). Computed tomography and magnetic resonance imaging are useful methods to determine localization and spread of ENA. ENA has less vascularisation, hence contrast retention is moderate or none compared to JNA (1,4,15). For its differential diagnosis, lobular capillary hemangioma (LCH or pyogenic granuloma), angiomatous polyps, neurofibromas and hemangioperistoma should be considered (16,17). LCH must be suspected in pregnant women. In the review study of el-Sayed, 7 of 12 LCH cases were originated from septum and one of these lesions was initially misdiagnosed as angiofibroma and according to the re-assessment results, diagnosis was corrected to LCH (17). In our case, LCH was diagnosed with the first biopsy and after the total excision, histopathology of tumor was reported as angiofibroma (Figure 1).

Tumor excision is the appropriate treatment option for angiofibromas. The role of preoperative embolization in ENA treatment is not clear. Somdas et al. reported posi-

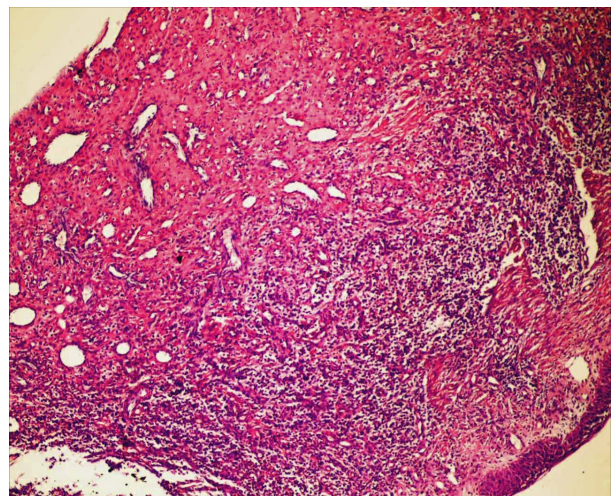


Figure 1. Reported histopathology result stated angiofibroma

Table1. Angiofibromas of septum

Authors	Year	Location	Age	Sex
Hiraide and Matsubara (6)	1984	R Nasal septum	13 year	M
Sarpa and Novelly (7)	1989	R Nasal septum	9 year	M
Bhargava et al. (8)	1995	R Nasal septum	24 year	M
Shah et al. (9)	2000	R Nasal septum	32 year	M
Handa et al. (10)	2001	L Nasal septum	8 year	M
Akbaş and Anadolu (1)	2003	R Nasal septum	50 year	F
		L Nasal septum	34 year	F
Somdas et al. (11)	2005	R Nasal septum	27 year	M
Castillo et al. (12)	2006	L Nasal septum	9 year	M
Tasca and Compadretti (4)	2008	R Nasal septum	57 year	F
Uyar et al. (13)	2009	L Nasal septum	19 year	M
Mohindra et al. (14)	2009	L Nasal septum	22 year	M
Our case	2009	R Nasal septum	22 year	M

tive results of preoperative embolization prior to excision of a septal angiofibroma (11). Castillo et al. reported an autoamputation of a septum-originated angiofibroma, for which operation was planned (12). As a result, ENA must be considered in the differential diagnosis of vascular tumors and it must be kept in mind that septum has a potential for these kinds of tumor localizations. It is supposed to be known that, clinic view of ENA is distinctive from JNA. The best treatment option is excision.

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